International Journal of Orofacial Myology and Myofunctional Therapy
Official Journal of the International Association of Orofacial Myology

Volume 45 | Number 1 | pp. 46-56 2019

Research Article

Investigating factors related to the acquisition of masticatory function in Down syndrome children

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Suggested Citation
Mizukami, M., et al. (2019). Investigating factors related to the acquisition of masticatory function in Down syndrome children. *International Journal of Orofacial Myology, 45(1)*, 46-56.
DOI: https://doi.org/10.52010/ijom.2019.45.1.4

INVESTIGATING FACTORS RELATED TO THE ACQUISITION OF MASTICATORY FUNCTION IN DOWN SYNDROME CHILDREN

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ABSTRACT
Down syndrome (DS) has the highest prevalence of any chromosomal abnormality identified in newborns. DS children have specific eating and swallowing difficulties such as poor tongue control, mouth opening, swallowing food without chewing, and both facial and occlusal abnormalities. DS children are also at high risk of aspiration, and swallowing food without chewing is considered to be a factor associated with increased risk of aspiration and eating problems. This study aimed to identify factors preventing the acquisition of masticatory function in DS children. The subjects were 75 outpatient DS children (44 males, age range 12 to 36 month-old, mean age 33.0 ± 7.0 month-old; 31 females, age 12 to 36 month-old, mean age 20.8 ± 8.0 month-old), who had not yet acquired masticatory function, out of 319 who visited the clinic between October 2012 and October 2017. The information necessary for assessment was retrospectively extracted from the medical records of the subjects. The items examined included age, birth weight, nutritional intake, picky eating, tactile hyperesthesia, cognitive development assessed by Ohta stage, gross motor function, occlusal condition by Hellman's dental age, and tongue thrust/lip closure/mastication while eating. The relationships between the acquisition of masticatory function and these items were investigated after one year of rehabilitation. The revealed age, low birth weight, picky eating, and gross motor function to be relevant factors. Among these, gross motor function was found to be the factor most strongly associated with acquisition of masticatory function.

KEYWORDS: Down Syndrome, mastication, tongue habits, low birth weight, eating, children

CONFLICT OF INTEREST
The authors have stated explicitly that there are no conflicts of interest in connection with this article. This study was funded by a Grant-in-Aid for Scientific Research (16K11870).

INTRODUCTION
Down syndrome (DS) is the most common chromosomal disorder in live births, with a prevalence rate of 53% (Wellesley, Dolk, Boyd, Greenlees, Haeusler, Nelen, Garne, Khoshnood, Doray, Rissmann, Mullaney, Calzolari, Bakker, Salvador, Addor, Draper, Rankin, Tucker, 2012). Many DS children have various eating and swallowing problems arising from DS-specific muscle hypotension, including poor tongue control, mouth opening, facial, and occlusal abnormalities, and susceptibility to fatigue (Hunter, 2005; Field, Garland, Williams, 2003; Cooper-Brown, Copeland, Dailey, Downey, Petersen, Stimson, Van Dyke, 2008; Van Dyke, Lang, Heide, Duyne, Soucek, 1990; Smith, Teo, Simpson, 2013). They also have a high risk of aspiration, and even silent aspiration has been reported (Field, *et al.*, 2003; Tutor, Gosa, 2012; Jackson, Maybee, Moran, Wolter-Warmerdam, Hickey, 2016). One of the factors raising the risk associated with eating is swallowing food without chewing. There are many studies of masticatory function of DS children (Hennequin, Allison, Faulks, Orliaguet, Feine, 2005; Rahul, Mathew, Jose, Thomas, Noushad, Feroz, 2015; Giannasi, Dutra, Tenguan, Mancilha, Silvam Fillietaz-Bacigalupo, Da Silva, Politti, Nacif, De Oliveira, Da Rocha, Romerom De Oliveira, De Mello Rode, Koga-Ito, Amorim, Salgado, Gomes, 2019). DS children have difficulties in acquiring masticatory function due to abnormal orofacial morphology and uncoordinated movements of the lips, tongue, and cheeks, as noted above (Faulks, Veyrune, Hennequin, 2002;

Hennequin, Mazillem Cousson, Nicolas, 2015). On the other hand, children with normal development children usually acquire chewing movement around 9-11 months of age and have well controlled jaw movement (Morris & Klein, 2000). whose development is normal usually acquire masticatory function at around 12 months of age with stabilization of mandibular movements.

Since many DS children have good interpersonal and social skills, they become increasingly independent as they mature and eventually become members of society. However, the eating problems of DS children with dysphagia can be an obstacle to societal participation. For instance, in most Japanese schools, intellectually impaired DS children have fewer opportunities to receive meals prepared for dysphagic children than those with severe motor and intellectual disabilities. Acquiring masticatory function is thus important for DS children to engage in society in addition to preventing choking and aspiration.

Previous studies on DS children were limited to treatment of mouth opening or tongue thrust, or focused mainly on dentition/occlusion (Faulks, *et al.*, 2002; Svensson, Romaniello, Wang, Arendt-Nielsen, Sessle, 2006; Faulks, Collado, Mazille, Veyrune, Hennequin, 2008). It is felt by the authors that there have been no reports on acquisition of masticatory function by young children with DS. This study aimed to identify factors preventing the acquisition of masticatory function in DS children ranging in age from 12 months (by this age, masticatory function has usually been acquired in children with typical development) to 36 months.

METHODS

Subjects

The subjects were 319 DS children (mean age at initial examination: 45.6 ± 75.6 months) out of 674 outpatients who visited an oral rehabilitation clinic for continued pediatric dysphagia rehabilitation between October 2012 and October 2017.

Study method

Data on the subjects were retrospectively extracted from the medical records. The evaluation results of children's feeding and swallowing functions included in these records were obtained through observation of actual eating behaviors involving food that had been brought from the children's homes on the occasion of their visits by dentists and dental hygienists who had at least 15 years of experience in pediatric dysphagia rehabilitation. Items extracted from these records were as follows: age, birth weight, nutritional intake, picky eating, tactile hyperesthesia, cognitive development assessed by Ohta stage (Hashino, Iida, Iwasaki, Ito, Sakiyama, Kitera, Matsumoto, Tsujimoto, Ikawa, Kishimoto T*,* 1997), gross motor function, occlusal condition by Hellman's dental age (Milo, 1932), and tongue thrust/lip closure/mastication while eating.

Age and birth weight

The corrected age in months was used for assessing the children that had been born prematurely. Children 12-36 months of age were included in this study and were divided into two groups: the "12-23 months" group and the "24-36 months" group. Subjects with birth weights ≥ 2,500 g were included in the "normal birth weight" group, and those weighing < 2,500 g at birth in the "low birth weight" group (Baumgartner, 1962).

Nutritional intake

Severe DS children with complete tube feeding were excluded. Other subjects were divided into two groups: the "partial tube feeding" group and the "oral intake only" group.

Evaluation of picky eating/tactile hyperesthesia

Picky eating (Rowell & Mcglothlin, 2015) was evaluated based on interviews with the DS children's parents or guardians. Subjects who ate only a specific food, adhered to limited types of foods over a long period, refused certain foods (meats/vegetables) or refused foods they had not previously eaten, were included in the "picky eating" group. Others were included in the "no picky eating" group.

Tactile Hyperesthesia (Kamide, Hashimoto, Miyamura, Honda*,* 2015) was evaluated by a dentist or dental hygienist. The evaluator slowly touched a DS children's cheeks first, and then the lips and gums with his/her palm or finger, and evaluated tactile hyperesthesia while observing apparent changes such as altered facial expressions, onset of weeping, and/or muscle hypertension. DS children who showed responses indicative of tactile hyperesthesia at any of these sites were included in the "tactile hyperesthesia" group, others in the "no tactile hyperesthesia" group.

Evaluation of cognitive development

Cognitive development levels were assessed based on Ohta stage (Hashino *et al.,* 1997). Hashino et.al. reported problems with eating, gross motor and emotional behaviors to be closely associated with low cognitive development levels (Hashino, et.al., 1997). Furthermore, they reported that these problematic behaviors were observed in the sensorimotor stage (Stage I) and were rarely found in stages at which understanding of symbols starts (Stage II or later). The cognitive functions were assessed by dentists, dental hygienists and speech therapists at the time of the initial examination and approximately one year later. The subjects were divided into two groups according to their cognitive development level: "Stage I", or sensorimotor stage (corresponding to the normal cognitive development level of children 18 months of age) and "Stage II or higher", or the initial stage of understanding symbols (corresponding to the cognitive development level of children 1.5-2 years old).

Evaluation of gross motor function

Gross motor function identified by GMFM (Russell, Palisano, Walter, Rosenbaum, Gemus, Gowlanad, Galuppi, Lane, 1998; Russell, Rosenbaum, Avery, Lane, 2002) was evaluated based on interviews with the parents or guardians and observation of DS subjects' behaviors. The results were classified into the following six levels: without steady head control, with steady head control, sitting up, pulling up to standing, aided walking, and unaided walking. Of these six levels, those in the categories from "without steady head control" to "pulling up to standing" were included in the "walking not acquired" group, while subjects in the "aided walking" and "unaided walking" categories were included in the "walking acquired" group.

Occlusal condition by Hellman's dental age

The number of teeth and occlusal condition were examined by dentists. The subjects were divided into two groups according to Hellman's dental age (Milo, 1932): "IA, IC", or incomplete deciduous dentition and unstable occlusion, and "IIA or higher", or complete deciduous dentition (Table 1).

Table 1. Classification of Hellman's Dental Age

I A	before eruption of deciduous tooth
I C	before attained of deciduous occlusion
II A	completion of deciduous tooth occlusion
II C	beginning of eruption of 1st molars and incisors
III A	completion of eruption of 1st molars and incisors
III B	exchanging of lateral dentitions
III C	beginning of eruption of second molars
IV A	completion of eruption of the second molars
IV C	beginning of eruption of third molars
VA	completion of eruption of third molars

Oral movement during eating

Evaluation of oral movement during eating was conducted using Showa University's Classification (Ayano, Tamura, Ohtsuka, Mukai, 2000).

Tongue thrust and lip closure during eating

Tongue thrust and lip closure were both evaluated during assisted feeding by parents to avoid possible influences of self-feeding. The subjects whose tongue protruded beyond the lower incisor teeth or lower lips were included in the "tongue thrust" group, and those whose tongue remained behind the lower incisors in the "no tongue thrust" group. For lip closure, the subjects with an elevated upper lip and those who were unable to close the lips, as well as the children who tried but failed to pull the upper lip downward, were included in the "no lip closure" group. The subjects who were occasionally or consistently able to close the lips were included in the "lip closure" group. Tongue thrust and lip closure were both evaluated on the following three occasions: when eating, during processing, and while swallowing.

Mastication

Mastication involves lateral tongue movement to position food for optimal chewing, and grinding food with the teeth and gums (Ayano, et.al, 2000; Morris, et.al.2000). The subjects who demonstrated these movements were included in the “masticatory function acquired” group and those who failed to demonstrate these movements in the “masticatory function not acquired” group. The acquisition of masticatory function was evaluated at the initial examination and after a one-year rehabilitation period.

Statistical analysis
Correlations of the acquisition of masticatory function at the one-year follow-up evaluation with items at the initial examination were examined using the Wilcoxon signed rank test. Furthermore, a logistic regression analysis was performed, with items showing a significance level of less than 10% as explanatory variables. The statistical significance level was set at less than 10%. All statistical analyses were performed using SPSS Version 23 for Windows (IBM Corp.).

Ethical considerations
This research was performed with approval from the ethics committee of the Nippon Dental University School of Life Dentistry (Approval number: NDU-T2018-25). Informed consent was obtained from the subjects and their parents or guardians through an opt-out system on the website of the Division of Rehabilitation for Speech and Swallowing Disorders, Nippon Dental University, Tama Oral Rehabilitation Clinic.

RESULTS

Subjects
Figure 1 shows the subject selection flow. Of the 674 patients who visited the pediatric dysphagia rehabilitation department during the aforementioned period, 319 had DS. Of these 319, we included 116 who were 12 to 36 months of age, and then excluded those who were completely reliant on tube feeding (1), visited the clinic less than five times in a year (8), and/or had acquired masticatory function (32) according to Ayano’s report (Ayano, Tamura, Ohtsuka, Mukai, 2000). Ultimately, 75 children (mean age at the initial examination: 20.9 ± 7.4 months) were included in this study. Subject characteristics are shown in Table 2. The subjects of 12-23 months of age group was 32 males and 22 females. The subjects of the 24-36 months of age group was 12 males and 9 females at the initial examination.

Table 2: Characteristics of subjects at the initial examination

Age (months) at the initial examination	Male[†] n (%)	Female[†] n (%)
12-23	32 (72.7)	22 (71.0)
24-36	12 (27.3)	9 (29.0)
Total	44	31

[†]Data consist of the number of participants (percentage by gender)

Correlations of masticatory function acquisition and general health status/eating and swallowing functions (Table 3)
By the Wilcoxon Signed Rank Test, there was one of the 75 subjects who had not yet acquired masticatory function at the initial examination, 38 (50.7%), who had acquired masticatory function, and 37 (49.3%) who did not acquire masticatory function after one year of rehabilitation. Sex, nutritional intake, tactile hyperesthesia and cognitive development level by Ohta stage showed no correlations with the

acquisition of masticatory function ($p < 0.10$). No correlation was observed between occlusal condition by Hellman's dental stage and tongue thrust/lip closure ($p < 0.10$). On the other hand, the number of children who had not yet acquired masticatory function was larger in the low birth-weight group than in the normal birth-weight group ($p = 0.078$). The proportion of subjects who had not yet acquired masticatory function was high among those with picky eating at the initial examination ($p = 0.054$). As for gross motor function, masticatory function was acquired by

a significantly higher proportion of "walking acquired" than "walking not acquired" subjects ($p = 0.002$). Logistic analysis revealed that only gross motor function correlated significantly with the presence/absence of acquisition of masticatory function ($p = 0.003$, odds ratio, 0.09; 95% confidence interval, 0.01-0.44).

DISCUSSION

As for eating and swallowing problems in DS children, Field et al. reported that the prevalence of oral motor dysfunction is 90% in children with cerebral palsy, followed by 80% in those with DS (Field, *et al.,* 2003). Jackson et al. performed videofluoroscopic swallow studies in 158 DS children, and found that 89 (56.3%) had frequent aspirations or penetrations (Jackson, *et al*., 2016). DS is reportedly associated with various causes when these children develop dysphagia, including DS-specific muscle hypotension, poor tongue control, facial and occlusal abnormalities such as micrognathia, mandibular protrusion, mouth opening, and oral dysesthesia (Field, *et al.*, 2003; Cooper-Brown, *et al.*, 2008; Faulks, *et al.*, 2008).

Considering the frequent aspirations or penetrations observed in DS children, treatment to improve dysphagia is urgently needed. This study was conducted to identify factors interrupting the acquisition of masticatory function, focusing on swallowing food without chewing, which is specific to DS patients. The subjects were limited to those requiring assistance with eating. This was done to exclude possible influences of inefficient self-managed eating behaviors on oral motor function (Morris & Klein, 2000; Tamura, Chigira, Ishii, Nishikata, Mukai, 2000).

The following 11 items were selected and examined as possible factors associated with masticatory function: age, birth weight, nutrition intake, picky eating, tactile hyperesthesia, cognitive development, gross motor function, occlusal condition and, tongue thrust/lip closure at rest, during processing and while swallowing. Of these 11 assumed items, nutrition intake, tactile hyperesthesia, cognitive development assessed by Ohta stage, occlusal condition by Hellman's dental age, tongue thrust and lip closure showed no correlations with masticatory function.

Children with severe motor and intellectual dysfunction are often tube fed, as oral intake is difficult due to reduced willingness to eat, impaired cognition and sensations arising from cerebral lesions or swallowing/respiratory disorders caused by brain stem lesions (Matsui, Suzuki, Hirayama, Kaga, Kurokawa, 1992).

These factors presumably contribute to difficulties in acquiring masticatory function. The subjects in this study, however, needed tube feeding to compensate for the lack of nutrients obtained with oral intake, rather than for dysphagia. This may account for the lack of a correlation between tube feeding and the acquisition of masticatory function.

No significant correlation was found between cognitive development levels by Ohta Stage (Hashino, et.al., 1997) and masticatory function in this current study. Stage I includes infants up to one and a half years of age, and 74 out of our 75 subjects were in Stage I in this study. Although the youngest subject was only 12 months old, the most plausible explanation of the failure to acquire masticatory function was the DS children's lower intellectual ability as compared to typically developing children. Further investigation of the involvement of cognitive development in older subjects is needed.

Table 3. Correlations between masticatory function and other characteristics

	Univariate			Multivariate	
	Masticatory function acquired (n =38)	Masticatory function not acquired (n =37)	*p-Value*	OR (95%CI)	*p-Value*
Age (months) *n* (%)					
12-23	24(63.2)	30(81.1)	0.070	-	-
24-36	14(36.8)	7(18.9)			
Sex *n* (%)					
Male	21(55.3)	23(62.2)	0.355		
Female	17(44.7)	14(37.8)			
Low birth weight					
Normal	32(84.2)	26(70.3)	0.078	-	-
Low Nutritional intake	5(13.2)	11(29.7)			
Nutritional intake					
Oral only	37(97.4)	34(91.9)	0.297		
Oral and tube feeding	1(2.6)	3(8.1)			
Picky eating					
No	38(100)	33(89.2)	0.054	-	-
Yes	0(0)	4(10.8)			
Tactile hyperesthesia					
No	38(100)	34(91.9)	0.115		
Yes	0(0)	3(8.1)			
Cognitive function (assessed by Ohta stage)					
Stage Ⅰ	37(97.4)	37(100)	0.507		
Stage Ⅱ or over	1(2.6)	0(0)			
Gross motor function					
Walking not acquired	22(57.9)	33(89.2)	0.002	0.08 (0.041-0.615)	0.003
Walking acquired	16(42.1)	4(10.8)			
Occlusal condition (Hellman's dental stage)					
ⅠA・ⅠC	30(78.9)	34(91.9)	0.104		
ⅡA or over	8(21.1)	3(8.1)			
T**ongue thrust when eating**					
No	31(81.6)	28(75.7)	0.366		
Yes	7(18.4)	9(24.3)			
Tonguethrustwhileprocessing					
No	17(44.7)	16(43.2)	0.541		
Yes	21(55.3)	21(56.8)			
Tonguethrustwhileswallowing					
No	24(63.2)	20(54.1)	0.286		
Yes	14(36.8)	17(45.9)			
Lip closing when eating					
Yes	21(55.3)	22(59.5)	0.447		
No	17(44.7)	15(40.5)			
Lip closing while processing					
Yes	5(13.2)	5(13.5)	0.615		
No	33(86.8)	32(86.5)			
Lip closing while swallowing					
Yes	17(44.7)	12(32.4)	0.196		
No	21(55.3)	25(67.6)			

†By Wilcoxon signed rank test § By Logistic regression analysis

Mastication involves tooth eruption in addition to gross motor functions, jaw control, and coordinated movements of the lips/cheeks/tongue. Morris & Klein (2000) reported that patients with unerupted teeth or insufficient chewing ability sometimes revert to munching as they are unable to maintain mastication. Therefore, we evaluated the occlusal condition by Hellman's dental age (Milo,1932), considering the impacts of tooth eruption status and jaw stability on mastication. In the current study, however, no correlation was found between occlusal condition and mastication. DS patients' masticatory functions are affected not only by oral problems such as muscle hypotension, mouth breathing and facial underdevelopment, but also by general health status including mouth breathing due to adenoidal and tonsillar diseases as well as gastrointestinal problems (Hennequin et.al., 2005; Faulks, *et al.* 2008; Hennequin et.al. 2015; Rahul, et.al. 2015; Giannasi, et/al/, 2019). Other studies (Allison, *et al*., 2004; Hennequin, Peyron, Faye, Hennequin, 2000) found relationships between occlusion and mastication in DS patients, but their patients were older than the subjects in the present study. Presumably because the subjects in this study were very young and developments of the jaw and teeth were incomplete, no clear correlation was observed. On the other hand, however, it has also been reported that the higher the age, the more difficulties the children have in acquiring mature masticatory function (Faulks, *et al*., 2008). It is considered to be important to provide swallowing rehabilitation to children with facial or oral underdevelopment.

In this study, lip closure and tongue thrust showed no correlation with mastication. Faulks demonstrated that DS patients are able to chew without acquiring lip closure or to swallow food by thrusting the tongue, and concluded that they are able to masticate even with incomplete lip closure (Faulks *et al*., 2008). However, another report found that lip closure to be an important factor in eating, while not being indispensable for acquiring masticatory function (Nakajima, Fujita, Asahi, 2012). Based on these findings, in DS patients, lip closure and tongue thrust are considered to have relatively small impacts on mastication. In children showing typical development, masticatory ability is not well developed at age one. At age two, this ability develops further with the eruption of milk molars. Similarly, in the DS children in our study, masticatory function improved with age.

Sixteen low birth-weight children (21.3%) were included among the subjects in this study. The proportion of children who had not yet acquired masticatory function was significantly higher in these 16 than in the other subjects with normal birth weight. According to some reports (Oliveria, de Lauzon-Guillain, Jones, Emmett, Moreira, Ramos, Charles, Lopes, 2015; Oudgenoeg-Paz, Mulder, Jongmans, van der Ham, Van der Stigchel, 2017; Stanley, Shepherd, Duvall, Jenkinson, Jalou, Givan, Steele, Davis, Bull, Watkins, Roper, 2018), low birth weight is associated with a higher risk of dysphagia, including delayed growth and motor/cognitive development, and oral tactile hyperesthesia. Terui et al. examined eating problems in low birth-weight children, and found mastication to be problematic, independently of picky eating and types of eating (Terui, Hiramoto, Arai, 2017). Morris & Klein (2000) reported that low birth-weight children's eating problems arise from immaturity of the sensory, nervous, organ and digestive systems. The low birth-weight children in our study accounted for 21.3% of participants. In these DS children, the proportion of subjects who had not yet acquired masticatory function was significantly higher in low birth-weight than normal birth-weight children, as demonstrated by the Kamide et al. study (Kamide, *et al*., 2015).

Children with picky eating tended to be less likely to acquire masticatory function than those without this feature. Problems such as oral sensitivity and selecting specific foods (Field, et al., 2003; Gisel, Lange, Niman, 1984) are factors which impede achieving improvements in the swallowing ability of DS patients.

In the current study, no significant correlation was found between tactile hyperesthesia and the acquisition of masticatory function. In performing rehabilitation, however, it is necessary to consider the possible linkage of tactile hyperesthesia and picky eating rather than considering these problems separately.

Among all factors evaluated in this study, gross motor function was found to be most strongly associated with the acquisition of masticatory function. Typically developing children start pulling themselves up to a standing position around 12 months of age, and acquire walking around age 15 months. Then, steady control of the jaw enables mastication to be maintained (Ayano et al., 2000). Accordingly, the subjects in this study were separated into two groups based on walking acquisition. However, 70 %

of the subjects were unable to pull themselves up to a standing position, even around age 12 months. In this study, development of gross motor function was delayed in DS children as compared to children developing normally, as reported previously (Hunter, 2005). This result showed stability and control of the lower limbs, reflecting maturation of motor functions, to be necessary for DS children, who tend to show delayed motor function development, to acquire masticatory function.

CONCLUSION

Among the various eating and swallowing problems experienced by DS children, the focus in the present study was on the acquisition of masticatory function and examined potentially relevant factors. It was determined that age, low birth weight, picky eating and poorly developed gross motor function correlated with the acquisition of masticatory function. Among these items, the most significant correlation was with gross motor function.

Acknowledgement

We express our deepest gratitude to the children with Down's syndrome and their families who participated in this study for their contributions to the present study.
We would like to thank my research partners and all individuals concerned for their generous cooperation.

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